



PARAPARESIS DUE TO HYDATID DISEASE OF THE THORACIC SPINE: A CASE REPORT

PARAPAREZİ İLE SEYREDEN TORAKAL OMURGA YERLEŞİMLİ KİST HİDATİK: OLGU SUNUMU

Turgut AKGÜL¹,
Hamza ARABAĞ²

¹Orthopedics and Traumatology Specialist, Şanlıurfa Training and Research Hospital, Şanlıurfa.
²Neurosurgery Specialist, Şanlıurfa Training and Research Hospital, Şanlıurfa.

SUMMARY:

Spinal hydatid disease is a rare pathology that often has a delayed diagnosis due to its atypical symptoms. Here, we present the case of a 63-year-old man with spinal hydatid disease. The patient presented with two months of progressive back pain and one week of progressive lower extremity weakness. His neurological examination showed progressive paraparesis of the lower extremity. The patient had a history of hydatid disease of the lung. Magnetic resonance images of the thoracic region showed an extradural vertebral and paraspinal multicystic lesion. Posterior decompression with laminectomy and debridement of all cystic material was immediately performed to prevent further neurological deficits. The pathological diagnosis was hydatid disease.

Key Words: Hydatid cyst, paraparesis, vertebral osteomyelitis.

Level of Evidence: Case report, Level IV

ÖZET:

Omurga kist hidatit hastalığı nadir görülmektedir ve atipik semptomları nedeni ile tanısı genellikle gecikmektedir. Torakal omurga yerleşimli kist hidatit hastalığı olan 63 yaşında erkek hasta çalışmamızda sunuldu. Hastanın iki aydır gittikçe artan sırt ağrıları, bir haftadır ilerleyen alt ekstremité güçsüzlüğü ve nörolojik muayenesinde ilerleyen alt ekstremité paraparezisi mevcut idi. Hastanın daha önceden akciğer kist hidatit hastalığı hikâyesi mevcut idi. Hastanın MR incelemesinde torakal bölge yerleşimli extradural ve paraspinal multikistik görünümü mevcut idi. Hastaya acil olarak nörolojik durumun kötüleşmesini engellemek için posterior dekompresyon ve debridman yapıldı. Kist hidatit tanısı, patolojik olarak da desteklendi.

Anahtar Kelimeler: Kist hidatit, paraparezi, vertebra osteomyelit

Kanıt Düzeyi: Olgu sunumu, Düzey IV

Address: Op. Dr. Turgut Akgül,
Ortopedi ve Travmatoloji Uzmanı,
Şanlıurfa Eğitim ve Araştırma
Hastanesi, Şanlıurfa.
Tel.: 0535 687 51 81
E-mail: doktorturgut@yahoo.com
Received: 22th July, 2012
Accepted: 1st September, 2012

CASE REPORT:

A 63-year-old man was referred to our hospital with back pain that had not resolved with medical therapy. The patient's initial clinical examination demonstrated back pain in the thoracic spinal region and an inability to walk 25 meters without support. The patient had a history of thoracotomy for hydatid disease of the lung seven years prior to his current presentation. At the hospital, the patient's clinical status worsened. His neurological examination showed progressive neurological deficits after one week of hospitalization. He became entirely unable to walk without support. His muscle strength was 3/5 in the bilateral lower extremities.

Plain radiography and magnetic resonance imaging (MRI) were performed while the patient's neurologic deficits were worsening. Blood samples were taken, and they revealed a slightly increased erythrocyte sedimentation rate (ESR) (30 mm/hr) and leukocyte count (16.0 million/ml).



Figure-1. Sagittal T2-weighted images of the thoracic region MRI showing a cystic lesion invading the spinal canal with spinal cord compression, collapse of T5 and T6 vertebral bodies, and intramedullary edema at the T4, T7 and T8 vertebral bodies.

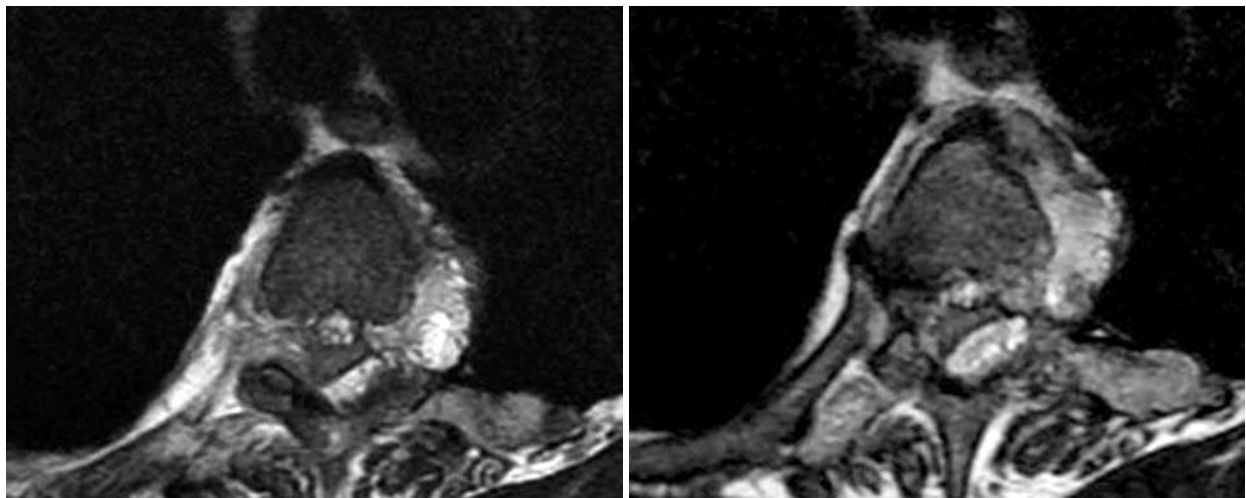


Figure-2. a,b. Axial MRI showing cystic lesion invading the spinal canal with spinal cord compression.

Plain radiographs of the thoracic spine showed compression of the vertebrae. MRI showed T5 and T6 vertebral height loss and T4 and T8 vertebral medullary edema. MRI investigation showed a mass composed of numerous round and oval shaped lesions with compression of the medulla spinalis. The lesion had intermediate T1 and high T2 signal intensities as well as faint rim enhancement after intravenous gadolinium (Figure-1,2).

An emergency operation was planned because of the patient's paraparesis. A posterior decompression with a T4-6 laminectomy and debridement of all infected structures was performed. Intraoperatively, yellow-white colored cyst material was found at the left pedicles of T4 and T5 (Figure-3).

The diagnosis of hydatid cyst disease was determined histopathologically. The patient was mobilized with a walker device on postoperative

day two. A postoperative MRI was performed and demonstrated decompression of the medulla spinalis (Figure-4).

The patient was referred to physical therapy after he was discharged from the hospital on postoperative day seven. Albendazole treatment was maintained for medical therapy. The patient was mobilized with a walker device, and had no other complaints three months postoperatively.

DISCUSSION:

Hydatid disease is most commonly found in the liver, spleen and lungs; musculoskeletal involvement is rarely seen^{5-6,10-11,15-16,19}. Spinal hydatidosis accounts for 1% of all cases of human hydatidosis. Spinal hydatid disease can be diagnosed with a combination of history, clinical examination, radiological evaluation and serological tests^{3-7,9-11,13-19}.

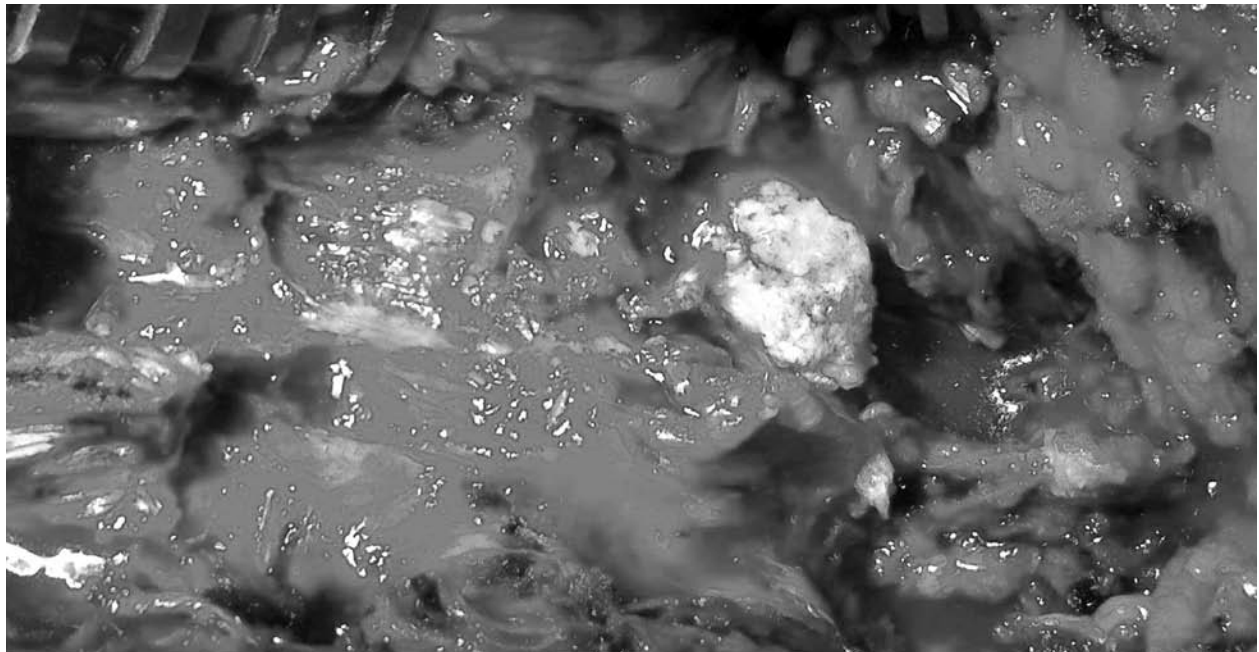


Figure-3. Perioperative photograph of the patient showing laminectomies and yellow-white coloured cyst material at the left pedicles of T4 and T5.

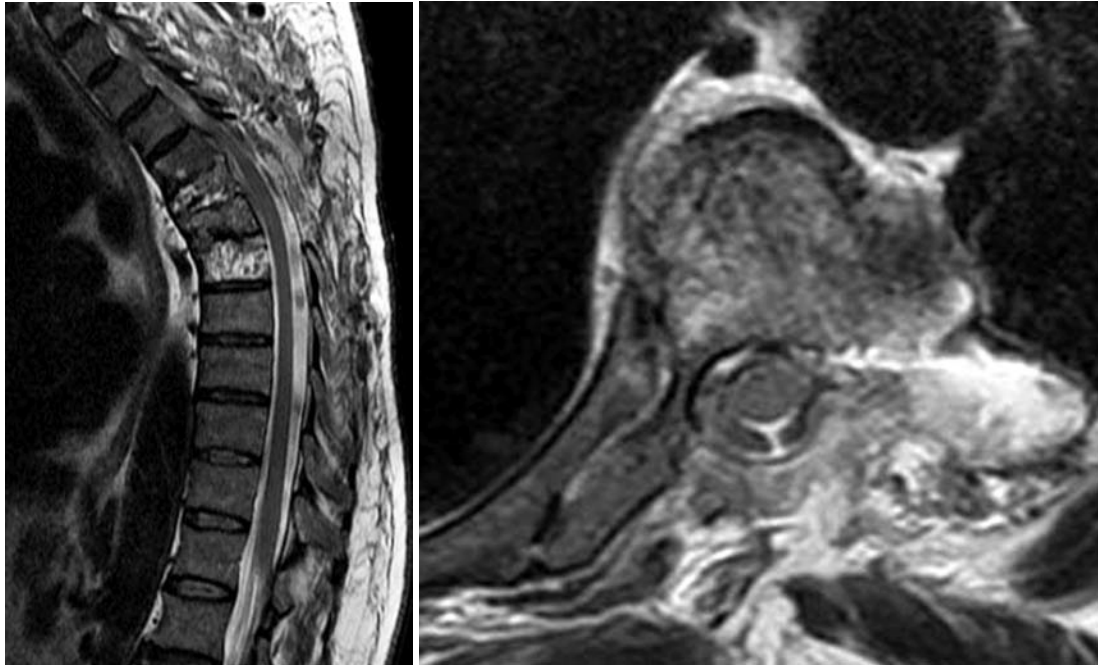


Figure-4. Postoperative (a) sagittal and (b) axial thoracic region MRI images show successful spinal canal decompression

A past history of hydatid disease is important in the diagnosis, because vertebral involvement frequently occurs due to the extension of pulmonary lesions; it rarely arises primarily from the vertebral body. In our case, the patient had a previous history of hydatid disease of the lung, for which he had received surgery and medical treatment. The hydatid cyst penetrates the vertebral corpus into the spongiosa that provides the least resistance^{4,6,9-10,14,16,19}. The cyst then grows multilocularly, causing slow destruction and expansion of the vertebrae. In later stages of the disease, the cyst breaks out of the vertebral body and invades the extradural space^{4,6,9-10,14,16,19}.

The clinical symptoms of hydatid disease are neither specific nor obvious. The clinical symptoms of spinal hydatid disease are related to cyst compression of the spinal cord, including pain and neurological deficits that may range

from segmental deficits to paraplegia^{4-7,9-11,13-19}. In our case, the patient had progressive neurological deficits.

Radiological evaluation may include plain radiography, CT and MRI. Plain radiographs during the initial stage of the disease show small osteolytic lesions with poorly-defined margins. At later stages, radiographs show multiple sharply demarcated osteolytic lesions or vertebral collapse, but these findings are not specific for hydatidosis^{2,3,5,8,11,12,15}. An MRI is useful for diagnosis and postoperative follow-up. Spinal hydatid cysts show typical features on an MRI. The cysts appear in clusters that resemble a bunch of grapes, and exhibit a hypointense rim that may enhance mildly after gadolinium administration^{3,8,12,14}. MRI shows vertebral expansion and destruction of the bony elements.

Braithwaite and Lees classified spinal hydatid disease into five groups using radiographic criteria. These groups are intramedullary, intradural extramedullary, extradural, vertebral, and paravertebral³. In our case, extradural, vertebral and paravertebral involvement were observed together.

Serological tests are useful to determine whether there is internal organ involvement, but are not useful in a diagnosis of bone hydatidosis^{17,19}.

The treatment of spinal hydatidosis involves a combination of medical and surgical treatment. The aim of surgical treatment is to remove the cyst without infecting the surrounding tissue, by a posterior, anterior, or combined approach^{4,6-7,9,13-14,17-18}. Generally, decompression through laminectomy and the removal of the cyst by a posterior approach is the initial surgery^{4,6,7,9,13,14,17,18}. In our case, we performed an immediate posterior spinal decompression and debridement of the cysts due to the patient's progressive neurological deficits. Anterior surgery was planned for the extensive removal of all the infected structures, but the patient refused this second surgery. In the literature, authors have suggested radical excision of all cysts with a combined anterior-posterior approach. Successful results have been reported with this treatment mode^{4,6-7,9,13-14,17-18}.

Systemic medical therapy alone was not enough for local control¹. Medical therapy is used to reduce the rate of local recurrence after radical resection¹. Mebendazole and albendazole are used for hydatid disease, albendazole has better intestinal absorption and a higher concentration within cystic material, making it a more effective treatment¹. In our case, albendazole was used for medical therapy.

Spinal hydatid disease is a rare pathology that is often misdiagnosed, or diagnosed late in the disease course, because of its atypical symptoms. Progressive neurological deficits are an indication for emergency treatment, which may include posterior decompression of the spinal cord. Medical therapy is useful for the control of local recurrence.

REFERENCES:

1. Arif SH, Shams-Ul-Bari, Wani NA, Zargar SA, Wani MA, Tabassum R. Albendazole as an adjuvant to the standart surgical management of hydatid cyst liver. *Int J Surg* 2008; 6: 448-451.
2. Beggs I, The radiology of hydatid disease. *Am J Roentgenol* 1985; 145: 639-648.
3. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. *Radiology* 1981; 140: 763-766.
4. Fares Y, Khazim R, El Zaatavi MM. Spinal hydatid disease and its neurological complications. *Scand J Infect Dis* 2003; 35: 394-396.
5. Gossios KJ, Kontoyiannis DS, Daskalogiannaki M, Gourtsoyiannis NC. Uncommon locations of hydatid disease. CT appearances. *Eur Radiol* 1997; 7: 1303-1308.
6. Günes M, Akdemir H, Tuğcu B, Günaldı Ö, Gümüş E, Akpınar A. Multiple intradural spinal hydatid disease: a case report and review of literature. *Spine* 2009; 34: E346-350.
7. Johannes LB, Folkert JVK, Olaf JWV, Paul IJMW. Long term follow-up of a patient with disseminated spinal hydatidosis. *Acta Orthop Belg* 2007; 73: 678-682.

-
8. Karantanas AH, Paterakis K, Karavelis A. Intervertebral disk hydatid cysts: MR imaging findings. *AJR Am J Roentgenol* 2003; 180: 1739-1740.
 9. Karray S, Zlitni M, Fowles JV, Douik M, Sliman N, Litaïem T. Vertebral hydatidosis and paraplegia. *J Bone Joint Surg* 1990; 72-B: 84-88.
 10. Keller TM, Schweitzer JS, Helfend LK, Chappell T. Treatment of progressive cervical spinal instability secondary to hydatid disease. A case report. *Spine* 1997; 22: 915-919.
 11. Kireşi DA, Karabacakoğlu A, Odev K, Karaköse S. Uncommon locations of hydatid cysts. *Acta Radiol* 2003; 44: 622-636.
 12. Mellado JM, Pérez del Palomar L, Camins A, et al. MR imaging of spinal infection: atypical features, interpretive pitfalls and potential mimickers. *Eur Radiol* 2004; 14: 1980-1989.
 13. Ozdemir HM, Ogun TC, Tasbas B. A lasting solution is hard to achieve in primary hydatid disease of the spine : long-term results and an overview. *Spine* 2004; 29: 932-937.
 14. Papakonstantinou O, Athanassopoulou A, Passomenos D, Kalogeropoulos I, Balanika A, Baltas C, Gouliamos A, Guglielmi G. Recurrent vertebral hydatid disease: spectrum of MR imaging features. *Singapore Med J* 2011; 52(6): 440.
 15. Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A. Hydatid disease from head to toe. *Radiographics* 2003; 23: 475-494.
 16. Rao S, Parikh S, Kerr R. Echinococcal infestation of the spine in North America. *Clin Orthop Relat Res* 1991; 271: 164-169.
 17. Salduz A, Koyuncu LO, Dıkıcı F, Talu U. Long term result of treatment for paraspinal and extradural hydatid cyst: case report. *Acta Orthop Traumatol Turc* 2009; 43: 267-271.
 18. Szypryt EP, Morris DL, Mulholland RC. Combined chemotherapy and surgery for hydatid bone disease. *J Bone Joint Surg* 1987; 69-B: 141-144.
 19. Turgut M. Hydatid disease of the spine: a survey study from Turkey. *Infection* 1997; 25: 221-226.